



Italian Journal of Gynæcology & Obstetrics

September 2022 - Vol. 34 - N. 3 - Quarterly - ISSN 2385 - 0868

Paraovarian serous cystadenofibroma: a rare case in a young woman and its management implications

Barbara Borelli ^{1,*}, Carlo Ticconi ¹, Rossana Telesca ², Benedetta Onelli ¹, Alessandro Mauriello ², Emilio Piccione ¹, Lodovico Patrizi ¹

¹ Department of Surgical Sciences, Section of Gynecology and Obstetrics, University of Rome Tor Vergata, Rome, Italy.

² Department of Systems Medicine, Section of Anatomic Pathology, University of Rome Tor Vergata, Rome, Italy.

ARTICLE INFO

History

Received: 01 May 2021

Received in revised form: 08 August 2021

Accepted: 03 December 2021

Available online: 12 September 2022

DOI: 10.36129/jog.2021.09

Key words

Ovarian/paraovarian cystadenofibroma; ovarian lesions in reproductive age; diagnosis and treatment; fertility preservation.

*Corresponding author: Barbara Borelli, Dr. Department of Surgical Sciences, Section of Gynecology and Obstetrics, University of Rome Tor Vergata, via Montpellier 1, 00133 Rome, Italy.
Email: b.borelli1990@gmail.com.
ORCID: 0000-0002-3439-5261.

ABSTRACT

Background. Cystadenofibroma of the ovary is a relatively rare benign tumor which originates from the germinal lining and the stroma of the ovary. Paraovarian serous cystadenofibromas are even rarer, with only three cases reported in literature. Since cystadenofibroma may present as a malignant tumor on preoperative examinations and/or macroscopically during surgery, it may cause both diagnostic and operative problems when treating young patients.

Case presentation. We present a case of a young patient with paraovarian serous cystadenofibroma, appearing as an absolutely benign lesion on preoperative ultrasound, but as a malignant tumor during surgery, that was finally treated by laparoscopy and completely preserving the patient's fertility.

Conclusions. The possibility of the occurrence of a paraovarian serous cystadenofibroma in a very young woman should be taken into account.

INTRODUCTION

Cystadenofibroma of the ovary is a relatively rare benign tumor which originates from the germinal lining and the stroma of the ovary [1]. Paraovarian serous cystadenofibromas are even rarer, with only three cases reported in literature [2-4].

This tumor may be solid, semisolid or cystic, depending on both the relative amount of the epithelial and stromal constituent, as well as on the secretory activity of the epithelial component [5]. Since cystadenofibroma may present as a malignant tumor on preoperative examinations and/or macroscopically during surgery, it may cause both

diagnostic and operative problems when treating young patients [6].

We present a case of a young patient with a paraovarian serous cystadenofibroma, which appeared as an absolutely benign lesion on preoperative ultrasound and as a malignant tumor during surgery. It was treated by laparoscopy and completely preserving the patient's fertility.

CASE PRESENTATION

An 18-year-old otherwise healthy woman was referred for a persistent and even growing ovarian cyst, which had been originally discovered about three months earlier, during a routine gynecological examination. The clinical examination was negative. Transvaginal ultrasound showed a normal uterus, with homogeneous myometrium; the endometrium was regular for thickness and echo structure; right ovary was normal for location and size, with a micropolycystic attitude; in the left ovary a non-vascularized anechoic cyst measuring 62 × 40 mm was detected (**Figure 1 A and A1**). Serum levels of the ovarian onco-markers (CA 125, CA 15.3, CA 19.9, AFP) were normal. Estroprogestinic therapy and close ultrasound checks were then prescribed.

The transvaginal ultrasound performed three months later revealed that the left adnexal non-vascularized anechoic cyst was unchanged, measuring 63 × 38 mm. After a collegial discussion of the case, surgical removal of the cyst was proposed to the girl. At laparoscopy, an approximately 65-mm diameter left paraovarian cyst, with smooth and regular external surface and citrin liquid content, was found. It was adherent to the left tube and contiguous with the upper pole of the ipsilateral ovary, which appeared perfectly normal; however, multiple millimeter papillae on the internal surface were unexpectedly found (**Figure B, C, D**). The cyst was completely removed by stripping technique, safeguarding both left tube and ovary (**Figure 1 E**). Due to the unexpected macroscopic appearance of the cyst, a frozen section was required. A serous cystadenofibroma was diagnosed.

The definitive histological examination showed a highly cellular and fibrous stroma, forming thick papillary projections, protruding within the cystic space, coated by a single layer of serous epithelium (**Figure 1 F and F1**), confirming the diagnosis of paraovarian serous cystadenofibroma.

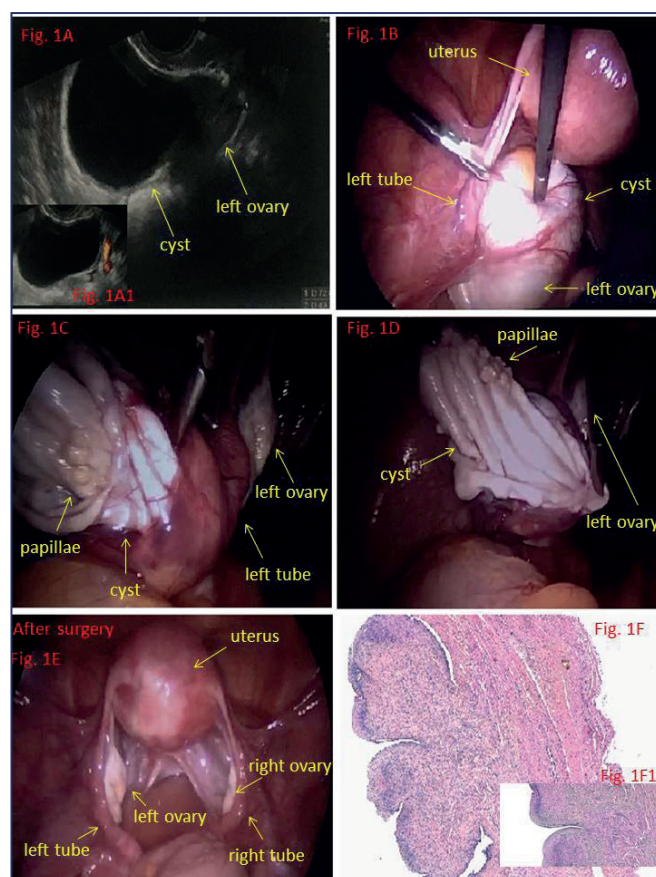


Figure 1. (A) Ultrasound aspect of paraovarian cystadenofibroma. (A1) Ultrasound examination with color doppler. (B) Macroscopic external surface of paraovarian cystadenofibroma. (C, D) Macroscopic internal surface of paraovarian cystadenofibroma, with papillae. (E) Anatomical appearance after surgery. (F) Microscopic feature of one of the papillary projection. It is constituted by highly cellular fibrous stroma and is covered by a single layer of flat epithelium (Haematoxylin-eosin stain. Original magnification 100x). (F1) At higher magnification, it is possible to appreciate the serous epithelium (Haematoxylin-eosin stain. Original magnification 200x).

DISCUSSION

Ovarian/paraovarian cystadenofibroma is a benign surface epithelial tumor which contains fibrous components. According to the epithelial cell type, it is classified into serous, endometrioid, mucinous, clear cell, and mixed categories. The serous type is the most common, representing 75% of all forms. The incidence peak of this tumor is between 40 and 50 years, and it is extremely rare at younger ages [1, 2]. It is often diagnosed preoperatively as a malignancy because of a solid component or irregular thick septa. It is usually cystic and thin walled, containing septation in 30.4% and solid nodules in 56.5-80% of cases [7, 8]. The most frequent appearance is a unilocular, complex cystic tumor (43.4%). The usefulness of color Doppler sonography in order to evaluate the vascularity of cystic wall,

septum and/or solid component has also been reported [7].

In this case, it preoperatively appeared as an absolutely benign tumor, with ultrasound features of a unilocular, thin walled cyst, without any septum or solid component, non-vascularized on color Doppler examination. It was only during laparoscopy that the problem of differential diagnosis with malignancy or borderline malignancy was posed. The frozen section was decisive in determining the type of surgical intervention, as well as in preserving the young patient's fertility.

CONCLUSIONS

As demonstrated by the clinical case we have reported, even though it is extremely rare, the possibility of the occurrence of a paraovarian serous cystadenofibroma in a very young woman should be taken into account.

COMPLIANCE WITH ETHICAL STANDARDS

Authors contribution

B.B.: Data curation, conceptualization, writing, review, editing. C.T.: Validation. R.T.: Software, writing. B.O.: Data curation, Software. A.M, E.P.: Supervision. L.P.: Resources, data curation, conceptualization, validation.

Funding

None.

Study registration

N/A.

Disclosure of interests

The authors declare that they have no conflict of interests.

Ethical approval

The scientific work was carried out in accordance with the ethical standards established in the Declaration of Helsinki.

Informed consent

Written informed consent was obtained from the patient for anonymized publication of this case report and accompanying images.

Data sharing

Data are available under reasonable request to the corresponding author.

REFERENCES

1. Cho SM, Byun JY, Rha SE, Jung SE, Park GS, Kim BK, et al. CT and MRI findings of cystadenofibromas of the ovary. *Eur Radiol.* 2004;14(5):798-804. doi: 10.1007/s00330-003-2060-z.
2. Czernobilsky B, Borenstein R, Lancet M. Cystadenofibroma of the ovary. A clinicopathologic study of 34 cases and comparison with serous cystadenoma. *Cancer.* 1974;34(6):1971-81. doi: 10.1002/1097-0142(197412)34:6<1971::aid-cn-cr2820340616>3.0.co;2-n.
3. Garikapati K, Kota LN, Premalatha P, Melam J. A rare case presentation of papillary serous cystadenofibroma in paraovarian cyst. *Int J Sc Research.* 2015;4(5):1889-91. Available at: <https://www.ijsr.net/archive/v4i5/SUB154643.pdf>.
4. Miura R, Fukui A, Funamizu A, Shigeto T, Yokoyama Y, Mizunuma H. A case of paraovarian serous cystadenofibroma that showed a malignant potential in image inspection. *Gyn Min Inv Ther.* 2016;5:96-8. doi:10.1016/j.gmit.2015.12.004.
5. Wolfe SA, Seckinger DL. Varied anatomical types of ovarian adenofibroma. *Am J Obstet Gynecol.* 1967;99(1):121-5. doi: 10.1016/S0002-9378(16)34500-8.
6. Groutz A, Wolman I, Wolf Y, Luxman D, Sagi J, Jaffa AJ, et al. Cystadenofibroma of the ovary in young women. *Eur J Obstet Gynecol Reprod Biol.* 1994;54(2):137-9. doi: 10.1016/0028-2243(94)90253-4.
7. Alcazar JL, Errasti T, Minguez JA, Galan MJ, Garcia-Manero M, Ceamanos C. Sonographic features of ovarian cystadenofibromas: spectrum of findings. *J Ultrasound Med.* 2001;20(8):915-9. doi: 10.7863/jum.2001.20.8.915.
8. Atri M, Nazarnia S, Bret PM, Aldis AE, Kintzen G, Reinhold C. Endovaginal sonographic appearance of benign ovarian masses. *Radiographics.* 1994;14(4):747-60; discussion 761-2. doi: 10.1148/radiographics.14.4.7938766.